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Rapidly progressive multifocal phyllodes tumour of the breast: A case report and review of the literature

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ABSTRACT

INTRODUCTION: Malignant transformation of a phyllodes tumour is a rare form of breast cancer, accounting for just 0.5% of all breast cancer cases.[1]
PRESENTATION OF CASE: We report a case of a 49 year old female with rapidly progressive, multifocal disease. She initially presented with two giant fibroadenomas which were excised. She represented eight months post surgery with two new lesions in the same breast, one suspicious, one suggestive of fibroadenoma. Biopsy was borderline. Surgery was therefore scheduled for wide local excision. At localisation two weeks later, at least eight lesions were seen on ultrasound scan. Three were removed as histology was at this point unknown to conserve the breast. Histology revealed intermediate grade DCIS, benign Phyllodes and borderline/malignant phyllodes. She was scheduled for mastectomy and immediate Strattice reconstruction. An MRI was performed pre-operatively to ascertain extent of disease. Two weeks post localisation, 13 lesions were identified. The right breast was entirely unaffected. Surgery interval was three weeks and final histology revealed 18 lesions, ranging from fibroadenoma through to borderline/malignant phyllodes with an incidental papilloma.
DISCUSSION: This is the first report of such rapid progression of disease, with 16 new lesions, of varied histology, developing in just an eight week period.
CONCLUSION: This case highlights the difficulty of forming a clear diagnostic and therapeutic pathway in this highly variable disease. Arguments for over and under treating these patients remain but those with any borderline/malignant potential have to be removed as recurrence is both common and aggressive, with a clear surgical margin the only proven protective factor.

1. Introduction

Malignant transformation of a phyllodes tumour is a rare form of breast cancer, accounting for just 0.5% of all breast cancer cases.[1] It was first described as early as 1774 but first fully classified in 1838 by Johannes Muller. It is a tumour of fibroepithelial cells with classically deep, leaf-like stromal fronds from which its name is derived. The histological classification is determined by a range of characteristics including stromal atypia, stromal overgrowth, mitotic count and tumour margins. Based on these criteria there is a spectrum of benign, borderline, borderline/malignant and malignant disease.[2]

Diagnosis is often difficult as the history and clinical findings often mimic that of the common benign presentation of fibroadenoma. Tumours most commonly occur in isolation and unilaterally, as do fibroadenomas. Reports of multifocal disease are rare and bilateral disease is also unusual with two previous reported cases of multifocality in the literature.[3] Features suggestive of phyllodes are: older presentation (phyllodes is most common in 45–50 year old women), a large mass (phyllodes tending to be larger than fibroadenomas, up to 30 cm being commonly reported in the literature), and an increase in size of a previously stable mass.

Pre-operative diagnosis is further hindered by the inaccuracy of imaging and tissue sampling in phyllodes with figures published for MRI only achieving diagnosis in 30% of cases and core biopsy providing a false negative rate in 39% of cases.[4,5]

In this case we highlight some of the issues surrounding diagnosis and classification through an unusual case of unilateral, multifocal, multi-grade phyllodes tumour.

2. Case presentation

A 49 year old initially presented with two symptomatic lumps in her left breast. They had been present for some years but had now grown in size. Mammogram revealed two, 6 cm lesions of benign appearance. Ultrasound also appeared consistent with fibroadenoma. She had no other breast problems or family history and had no known medical conditions. Excision biopsy took place and final histology was that of fibroadenoma and she was discharged. When reviewed retrospectively, the original diagnosis of fibroadenoma

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with no atypical features was confirmed, and the changes were noted to extend to the margins of both nodules.

Eight months post op, she re-presented with a new palpable lump in her left breast confirmed on mammogram and ultrasound as a 25 mm lesion with benign appearance in the upper outer quadrant. Incidentally another lesion was identified in the lower inner quadrant, 12 mm of benign appearance. Core biopsy of the larger lesion revealed benign phyllodes tumour, strongly ER and PR positive. She was scheduled for wide local excision.

At localisation, only two weeks later, eight lesions were now visible on ultrasound, mainly clustered in the subareolar region and laterally. To conserve maximal breast and with pathology unknown, three wide local excisions were performed. Histology revealed benign phyllodes tumour, intermediate grade DCIS and borderline/malignant phyllodes.

This lady was discussed at the Multidisciplinary Team Meeting and in view of her multifocal disease with rapid progression and borderline/malignant histology she was scheduled for mastectomy with immediate sub pectoral implant and Strattice reconstruction for three weeks' time.

In view of the rapid development of the lesions an MRI was performed four weeks post op to ascertain extent of disease which showed further increase in nodularity with thirteen lesions now visible, none with malignant features, though histology was already suggestive of this.

Her surgery was performed two weeks later without complications and final histology revealed 18 nodules, ranging from 6 to 22 mm in size. The final diagnosis was multiple phyllodes tumours showing low-grade malignant or borderline features and a single intraductal papilloma. There was moderate atypia. No stromal overgrowth was identified. The highest mitotic count was 9 per ten high power fields. A mastectomy was performed and all margins were free of tumour.

3. Discussion

This case highlights the huge variation in behaviour of this tumour, which even in isolation, with clear surgical margins is prone to recurrence in 30% of cases with borderline histology, and 65% in malignant cases. In 70% of cases the recurrence is also at a higher grade than the original tumour, again emphasising the importance of appropriate diagnosis and resection of this rare but difficult to manage tumour.[6]

The key element in terms of risk reduction for recurrence is achieving a clear surgical margin, which is often not the case given the working diagnosis of giant fibroadenoma, often supported by a benign core biopsy in which a simple capsular excision, not wide local excision is performed. Additional information from core biopsy immunohistochemistry is currently unsupported and so diagnosis remains a challenge.[7,8] In all cases where phyllodes is confirmed and clear margins not achieved, surgical re-excision is indicated.

Regarding adjuvant therapy, there is an argument for radiotherapy, though not commonly practiced it does appear to reduce both local recurrence and metastasis from this disease.[9] Currently there is no substantial evidence to support chemotherapy for phyllodes tumours.[10,11]

4. Conclusion

Phyllodes tumour encompasses a large spectrum of disease, from benign, quiescent disease to malignant tumours with a high propensity for both recurrence and distant metastases. The patterns of disease are hugely varied as demonstrated by this case and many others,[12] making clinical diagnosis exceedingly difficult.

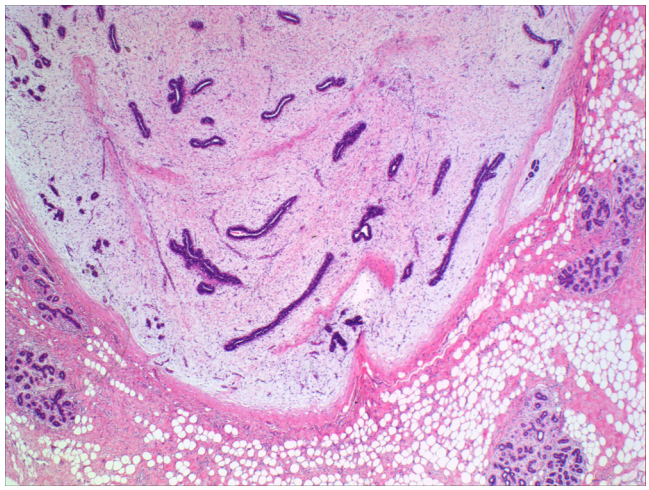

Fig. 1. Benign specimen 20× magnification.

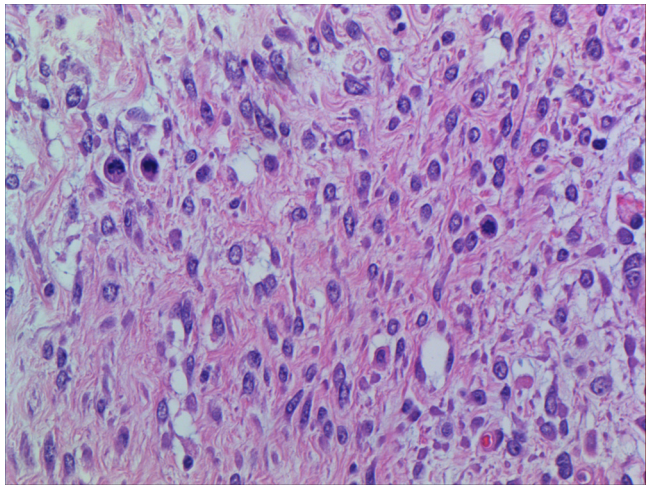

Fig. 2. Microscopic specimen: borderline-Malignant 400× magnification.

If large benign feeling lumps have to be excluded as phyllodes then this would inevitably lead to a vast increase in number of core biopsies required, which themselves appear flawed in detecting phyllodes tumours. Diagnosis is difficult and shows no sign of becoming easier based on early studies into further imaging

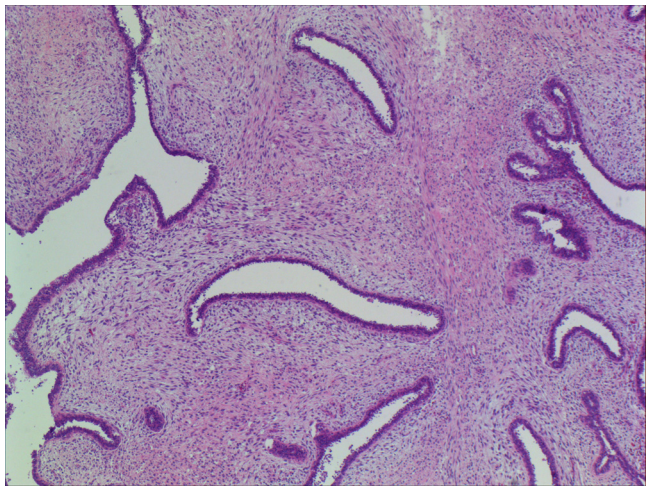

Fig. 3. Microscopic specimen: borderline-malignant 40× magnification.

Fig. 4. Macroscopic sections of mastectomy sample of left breast showing extent and variation of tumours.

and immunohistochemistry.[13] Though classification is very subjective based on which criteria are used, the fundamental point is that even in benign disease, an adequate, clear surgical margin must be achieved to limit local and distant recurrence of disease as metastatic disease has no definitive treatment at this point in time (Figs. 1–4).

Conflict of interest statement

No conflict of interest declared. No sources of funding to declare.

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Ethical approval

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Authors' contributions

D. White – study design, data collections, data analysis, writing, literature review.

T. Irvine – review of paper for publication. Corresponding consultant.

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